

## TUMORS VENTRAL TO THE SACRUM

LORIN D. WHITTAKER, M.D.\*

FELLOW IN SURGERY, THE MAYO FOUNDATION

AND

JOHN DEJ. PEMBERTON, M.D.

DIVISION OF SURGERY, THE MAYO CLINIC

ROCHESTER, MINN.

THE region ventral to the sacrum and sacrococcygeal joint is the site of many complex fetal changes. While tumors arising in this region are rare—according to Ewing<sup>3</sup> occurring only once in 34,582 tumors of the newborn, and according to statistics at The Mayo Clinic, for the period 1922–1936, occurring in only one out of approximately every 40,000 registrations—their incidence is such as to prompt consideration of their pathogenesis, diagnosis and treatment.

This study is based on a series of 22 cases of tumor ventral to the sacrum seen at The Mayo Clinic from 1922 to 1936, inclusive, in which the diagnosis was confirmed at operation. Hundling<sup>8</sup> has previously reported 19 such cases which were encountered at the clinic prior to 1922, and Fletcher, Woltman and Adson<sup>5</sup> have previously reported in detail the five cases of chordoma which are included in this report.

Located embryonically in the ventral sacrococcygeal region are the spinal cord, nerve tissue, postanal portion of the hindgut, neurenteric canal, caudal end of the notochord and the bony vertebral canal. The embryonic development<sup>2</sup> and subsequent disappearance of the structures in this region aid in explaining the origin of these tumors. The primitive streak lies posterior to the neural groove and arises, as does the neural groove, from the embryonic area. The notochord is formed by the proliferation of cells from the anterior end of the primitive streak. It is epithelial in origin. Early in its development the notochord is surrounded by secondary mesoderm. This, then, becomes the primitive vertebral column. As the vertebral bodies are formed, after the fourth week, the continuity of the notochord is broken. The notochord disappears in the region of the vertebral bodies, but persists in the region of the intervertebral fibrocartilages. After the fourth month the notochord has disappeared as a separate structure, but remnants are believed to persist as the nucleus pulposus of the intervertebral fibrocartilages. Linck and Warstat<sup>9</sup> were of the opinion that rests of chordal cells exist outside the axial skeleton anlage, and these can be recognized in the sacrococcygeal region in the adult. These rests may lie either in the body of the sacral vertebrae or on their anterior or posterior aspects.

As the notochord is forming, the primitive groove appears on the surface of the primitive streak. The anterior end of the groove, which is embraced by the posterior portion of the neural fold, deepens and forms a canal between the neural groove and the ventral-lying entodermal vesicle, which is the primi-

---

\* Now residing in Peoria, Illinois. Submitted for publication May 14, 1937.

tive intestinal canal. This is called the "neurenteric canal" (Fig. 1), and it gradually moves to a position caudal to the proctodeum. The neurenteric canal is but a transitory passage and disappears in man before the neural groove closes to form the neural tube.

The postanal gut is formed by the union of the proctodeum and that portion of the hindgut proximal to the neurenteric canal. The proctodeum is a surface depression limited at its depth by the proctodeal membrane. The disappearance of this membrane completes the formation of the rectum and anus. The union of proctodeum and hindgut, however, does not take place at the distal end of the hindgut.<sup>2, 10</sup> That portion of the hindgut distal to the union with the proctodeum is known as the "postanal gut," and it lies on the ventral

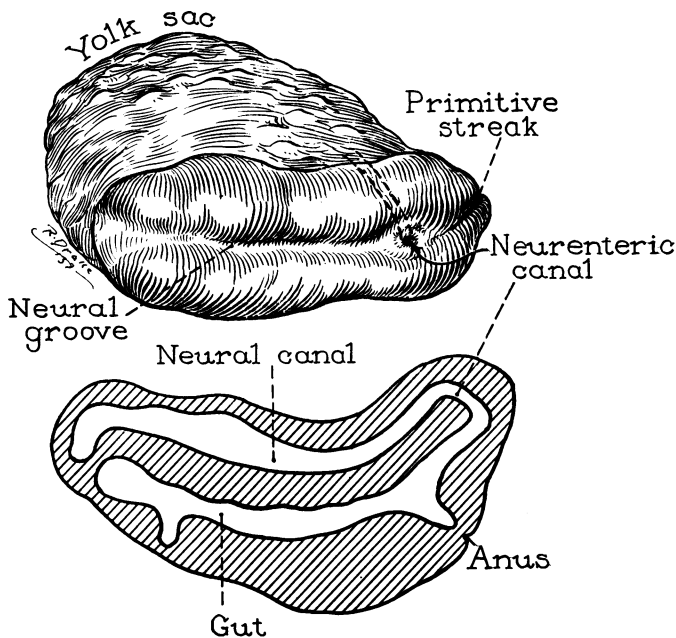


FIG. 1.—Dorsal view of a human embryo 1.54 Mm. long (modified after Graf Spee). Longitudinal (sagittal) section through an advanced embryo of Bombinator (modified after Goetta in Herturg, Oscar, and Mark, E. L.: Textbook of the Embryology of Man and Mammals. London, Swan, Sonnenschun and Co., 1892, p. 120, fig. 80).

surface of the coccyx (Fig. 2). This structure atrophies early. It is apparent, therefore, how these primitive structures, as they atrophy, may leave a nidus for further growth, with the formation of a tumor.

Tumors ventral to the sacrum have at times been popularly known as "Middeldorpf" tumors, but this term should be limited to those teratomatous tumors that arise from the postanal gut, as first clearly described by Middeldorpf.<sup>10</sup> Tumors ventral to the sacrum may vary from those the size of an egg to large growths which fill the true pelvis and push the anus and genitalia forward. They may be rather superficial or be quite deeply placed within the hollow of the sacrum. The tumors are usually encapsulated and rarely invade the rectum but, as will be shown later, they may invade the bony sacrum or encroach on the intervertebral foramina and compress the nerve trunks.

The general symptoms produced vary greatly with the associated pathologic changes. In some cases there may be no symptoms; or an indefinite dull ache or pain in the lower part of the back or pelvis, which may occasionally be associated with constipation, may be the only complaint. In other cases serious nerve encroachment may produce weakness in the legs, sciatic pain, rectal or urinary incontinence, and areal numbness. In some cases draining perirectal sinuses from incised dermoids are seen. Consequently, digital examination of the rectum is the most valued single procedure in the diagnosis of tumors ventral to the sacrum, and this alone almost always will give the clue to the diagnosis. It is important, however, to examine thoroughly the hollow of the sacrum. If this is done, the true diagnosis is established and

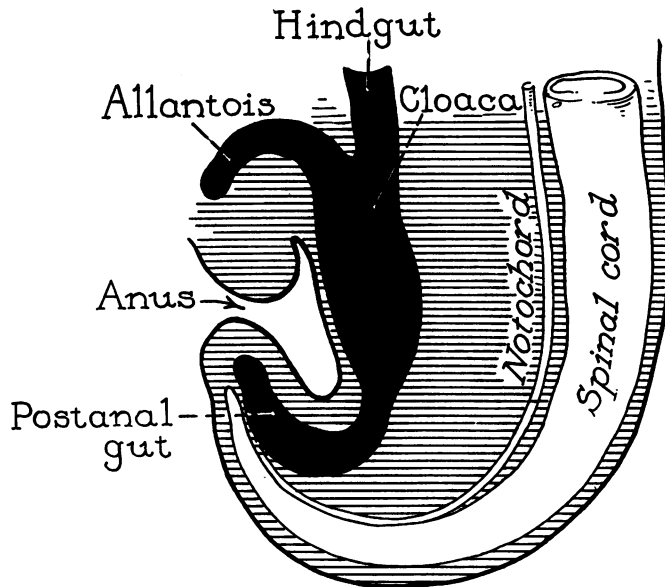


FIG. 2.—Reconstruction of the hindgut of an embryo 6.5 Mm. long (modified after Keibel).

the patient may be saved needless hemorrhoidectomy, prostatic massage, arthritic treatment, or even laminectomy, for relief of his symptoms.

Conditions to be considered in the differential diagnosis, suggested by this series of cases, include: Cord tumor, tumor of the cauda equina, anterior spina bifida with meningocele, primary sarcoma of the sacrum, pelvic tumors such as ovarian cysts, fibromyomata or adenomyomata, masses of metastatic lymph nodes, rectal fistulae and perirectal abscesses.

The treatment of choice is surgical removal, utilizing the posterior approach through a modified Kraske incision. Radium and roentgen therapy are advised when removal is incomplete, since prolongation of life and relief of symptoms can frequently be obtained by these measures (Table I).

**CHORDOMATA.**—The term "chordoma" was first used by Ribbert, in 1894, and the first sacrococcygeal chordoma was described by Feldmann,<sup>4</sup> in 1910. The term is now used to describe tumors whose cellular structure has the characteristic appearance microscopically of the notochord. There are approximately 125 cases of chordoma on record.<sup>12</sup>

TUMORS VENTRAL TO THE SACRUM

TABLE I  
TUMORS VENTRAL TO SACRUM

Diagnosis	No. of Cases	Sex		Age Range	Predominant Symptoms	No. of Cases Reveal- ing Mass Rectally	Prognosis
		M.	F.				
Chordoma...	8	5	3	¼ to 65	Pain in lower part of spine; nerve involvement;* constipation	8	Poor
Dermoid cyst.	9		9	⅓ to 48	Dull pelvic pain; draining sinus; mass at birth	7†	Good
Teratoma....	1		1	7	Perineal mass at birth	‡	Good
Fibrosarcoma.	1	1		51	Pain in rectum; nerve involvement	1	Poor
Chondromyxosarcoma	1	1		73	Mass; loss of weight; constipation	1	Poor
Squamous cell epithelioma	1	1		53	Sciatic pain	1	Poor
Fibroma.....	1		1	29	Sacral pain	1	Good
Total.....	22	8	14	3 mos. to 73 yrs.		19	

\* Sensory disturbance noted in three cases.

Numbness, rectal or urinary incontinence.

† No record of rectal examination in the cases of two infants.

‡ No record of rectal examination.

Chordomata arising from the remnant of the notochord may, as stated previously, have as their origin the nucleus pulposus or the rests of chordal cells outside the axial skeleton. However, since chordomata occur with about equal frequency in the sacrococcygeal and dorsum sellae regions, and since one has not been reported in the thoracic region, it seems more plausible to many that their origin is in the chordal cell rests outside the axial skeleton anlage. Anatomically the tumors seem to arise in relation to the vertebral body rather than the nucleus pulposus.

Sacroccygeal chordomata may arise within the vertebral body and project either ventrally or dorsally, or both; or they may arise ventral or dorsal to the sacrum. It is only in those arising or projecting ventrally, however, that we are interested. These tumors may grow to a large size, are usually encapsulated, may invade intervertebral foramina and cause pressure on nerve structures, and may cause marked bony destruction; they rarely if ever, however, involve the rectum. Metastasis is rarely seen, although the incidence of recurrence is quite high, being given by Stewart<sup>12</sup> as 86.5 per cent. Recurrent lesions are local and recurrence is usually due to incomplete removal of the original growth. Grossly these tumors are gelatinous, lobulated and usually quite friable; microscopically, certain criteria have been established

on which to base the diagnosis. Fletcher, Woltman, and Adson,<sup>5</sup> for example, have based the diagnosis on: "(a) The formation of intracellular and extracellular mucus; (b) the presence of physaliphorous or vacuolated mucus containing cells; (c) the lobulated arrangement of the tumor cells, which usually grow in cords; (d) the occasional occurrence of vacuolation of the nuclei; and (e) the close resemblance to notochordal tissue as seen in the nuclei pulposi of the intervertebral disks."

The last five cases in this group of cases of chordoma have been previously reported in detail by Fletcher, Woltman and Adson. They will be reviewed briefly here only to complete the series:

**Case 1.**—A girl, age five years, was brought to the clinic August 6, 1930. She had had increasing constipation for two and one-half years, and obstipation had been present for ten days. Rectal examination revealed a mass posterior to the rectum, pushing it anteriorly against the pubis. This mass measured 8x6x4 cm. and nearly filled the pelvis. Roentgenograms of the pelvis were negative. The mass was removed, as completely as possible, August 15, 1930. It extended from the ventral sacrum back around the coccyx and out into the right buttock. Radium was applied following operation. Five months later the bowels were regular and the patient was doing nicely. The microscopic report was chordoma.

**Case 2.**—A girl, age three months, was brought to the clinic March 11, 1935. In the previous three weeks her right hip had appeared larger. Constipation had also been present for three weeks. Examination revealed a firm mass in the right buttock. Rectally the mass was felt on the right, 6 cm. in diameter, pushing the rectum to the left. Roentgenograms of the pelvis were negative. The mass was incompletely removed March 13, 1935. It was very friable, vascular, and was invading the right gluteal muscles. The patient returned home and later died. The microscopic report was chordoma.

**Case 3.**—A man, age 39, registered at the clinic June 25, 1936. He had had pain in the lower part of his back and at the end of his spine for the previous five months. This pain was worse at night. During the same period an increasing constipation was noted. Rectal examination revealed a firm, smooth rounded mass, 7x5 cm., posterior to the rectum and attached to the sacrum. It was not tender. The lumen of the rectum was partially obstructed. Roentgenograms revealed destruction of the lower half of the sacrum. The tumor was removed as completely as possible July 7, 1936. It was encapsulated. Roentgen therapy was instituted following operation. On discharge the patient was free of symptoms and only some roughness of the sacrococcygeal area persisted. The microscopic report was chordoma (Fig. 3).

**Case 4.**—A woman, age 65, registered at the clinic September 4, 1923. She had had a severe, sharp pain at the end of her spine for five years. Hemorrhoidectomy and injection of the peri-anal nerves with alcohol, in 1920, had given no relief. There was sciatic projection of this pain, and rectal incontinence and some loss of vesical control was also noted. Rectal examination revealed a relaxed anal sphincter and a mass ventral to the sacrum. Sensory impairment of the fourth and fifth sacral nerves was found. Roentgenograms revealed destruction of the upper portion of the sacrum. On October 6, 1923, the tumor, which had invaded the sacrum, was incompletely removed and radium was applied. The patient died two years later. The microscopic report was chordoma.

**Case 5.**—A man, age 58, registered at the clinic January 26, 1925. He had had progressive tenderness and pain at the end of his spine for three months. Rectal examination revealed a hard round mass, 4 cm. in diameter, posterior to the rectum. It was not tender. Roentgenograms revealed necrosis of bone in the sacrum. The tumor was removed as thoroughly as possible January 30. Radiotherapy was then instituted. Nine years later the patient felt well. The microscopic report was chordoma.

**Case 6.**—A man, age 47, registered at the clinic for the second time April 6, 1925, with the complaint of pain and numbness in the region of his rectum and coccyx of 18

## TUMORS VENTRAL TO THE SACRUM

months' duration. He had also had progressive trouble with his bladder for one month. Rectal examination revealed a relaxed anal sphincter associated with anesthesia. A firm, smooth, fixed mass, 7 cm. in diameter, was palpated ventral to the sacrum, and roentgenograms revealed destruction of the lower two-thirds of the sacrum. On May 9, 1925, incomplete removal of the mass was accomplished. It was friable and resembled a colloid tumor. Roentgen therapy was administered. In 1929 a recurrent growth was partially removed. The patient died five years after the onset of his symptoms. The microscopic report was chordoma.

**Case 7.**—A man, age 61, registered at the clinic December 9, 1927. He had had progressive pain in his right hip associated with sensory disturbance for eight months. Urinary incontinence had been present for three weeks. Rectal examination revealed a relaxed sphincter and a firm mass filling the pelvis, apparently arising from the ventral aspect of the sacrum. Roentgenograms revealed some destruction of the sacrum.

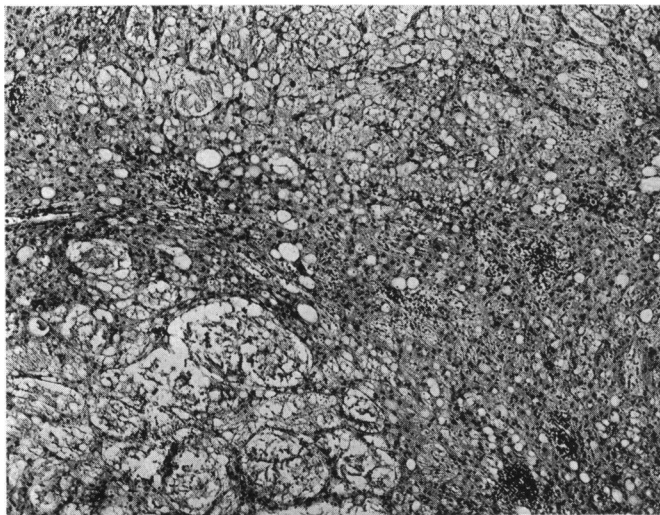


FIG. 3.—(Case 3). Photomicrograph of chordoma.

Biopsy only was attempted and radiotherapy was advised. The microscopic report was chordoma.

**Case 8.**—A man, age 26, first registered at the clinic August 19, 1930. He had had pain at the end of his spine, which was projected to the genitalia and inner thigh, and increasing constipation for one year. Rectal examination revealed a fixed tumor, 7.5 cm. in diameter, ventral to the sacrum. He was operated upon, elsewhere, and was given radium treatment. He returned to the clinic January 7, 1932, complaining of severe pain. Examination at this time revealed urinary and rectal incontinence and a mass ventral to the sacrum. Roentgenograms revealed cystic degeneration of the right half of the sacrum. The mass was removed as completely as possible January 14, 1932. On discharge the patient was free of pain and the function of his bladder had improved. The microscopic report was chordoma.

Five of these eight patients were males and three females. In only two cases was there any suggestive history of previous trauma. The ages ranged throughout the life span, two of the patients being children. Sacrococcygeal chordomata are quite rare in children.

Pain was the initial symptom in the case of each adult patient. This pain was usually localized in the back at the lower end of the spine. Sciatic projection was frequent. Later, sensory disturbances frequently appeared,

as did urinary and rectal incontinence. Involvement of nerves depended on encroachment of the tumor on the sacral nerve plexus and trunks. Constipation was frequently noted. Roentgenograms revealed some bony destruction of the sacrum in five of the cases. The duration of symptoms varied from three weeks to five years, the average being 17 months. Rectal examination revealed a mass in each case, and was the most valuable single finding in the examination. The mass was firm, usually smooth, fixed to the sacrum, and was not tender.

The diagnosis can be made positively only after microscopic examination. However, the presence of a firm, smooth, fixed and nontender mass ventral to the sacrum, associated with pain in the sacral region, later with sensory disturbances and usually with rectal and urinary incontinence, is quite suggestive of sacrococcygeal chordoma. The conditions which most frequently have to be considered in the differential diagnosis of sacrococcygeal chordoma are: Tumor of the cauda equina, meningocele, and sarcoma or enchondroma of the sacrum.

Surgical removal of the tumor as completely as possible offers the best chance for cure. The Kraske type of incision posterior to the rectum, with removal of the coccyx, probably gives the most satisfactory approach to these tumors; however, the dorsal approach, unroofing the tumor and removing it intrasacrally, may be a better approach when marked involvement of the sacrum is present. Because of its ramifications, complete removal of the tumor is sometimes very difficult.

The prognosis in this group of cases is quite poor and the percentage of local recurrence is extremely high. However, surgical removal followed by extensive irradiation does prolong life, relieve pain and often gives a long period of freedom from symptoms. Chesky<sup>1</sup> reported the average length of life after operation to be six and one-half years.

**DERMOID CYSTS.**—Dermoid cysts are ectodermal in origin and are lined with epithelium which is constantly secreting and desquamating. Such cysts are filled with sebaceous material and may contain ectodermal derivatives. These cysts occur where ectodermal structure is not found under normal conditions. It is generally agreed that such cysts originate by inclusion of a portion of ectoderm during faulty coalescence of cutaneous surfaces in embryonic life. They occur along the course of ectodermal invaginations. Those presenting as tumors ventral to the sacrum may arise from the proctodeal membrane or from coccygeal vestiges. Galletly<sup>6</sup> was of the opinion that they may also arise from the neurenteric canal, which is an ectodermal invagination.

Dermoid cysts may be located close to the sacrococcygeal region or be quite deeply situated in the hollow of the sacrum. If large, they may bulge into the perineum and push the genitalia forward or they may encroach on the rectal lumen. These cysts are encapsulated, have little or no blood supply of consequence, never invade the rectum, but if large may infrequently cause erosion of the sacrum and injury to nerve trunks. They are not fixed primarily.

ABBREVIATED REPORT OF NINE CASES OF DERMOID CYSTS

**Case 1.**—A woman, age 30, registered at the clinic November 15, 1922. She had undergone an operation for a perirectal "abscess" elsewhere, and a draining perirectal sinus had persisted. She had also been subjected to three operations for fistula-in-ano. Rectal examination at the clinic revealed a definite thickening on the posterior rectal wall 7.5 cm. above the anal margin. On August 31, 1923, the sinus tract was dissected out, and it was found to lead to a small cyst anterior to the sacrococcygeal joint. The pathologic diagnosis was dermoid cyst containing hair.

**Case 2.**—A woman, age 23, registered at the clinic June 2, 1925. She had had urinary difficulty for six months and partial rectal incontinence for five months. Rectal examination revealed a firm mass ventral to the sacrum which pushed the rectum to the right and displaced the pelvic organs upward. Roentgenograms of the pelvis were negative. On June 8, 1925, a large mass, 10 cm. in diameter, which was buried in the hollow of the sacrum, was completely removed. Complete relief of symptoms followed. The pathologic diagnosis was dermoid cyst containing only fat.

**Case 3.**—A girl, age four months, was brought to the clinic August 9, 1926. A tumor over her right buttock had been noticed at birth. It had recently become more prominent. Examination revealed a soft tumor with firm nodules extending up to the sacrum, and roentgenograms revealed a soft tissue shadow below the pelvis on the right. On August 14, 1926, a mass 10 cm. in diameter, arising ventral to the sacrum and extending down to the right buttock, was completely removed. Complete relief of symptoms followed. The pathologic diagnosis was polycystic dermoid containing fat and hair.

**Case 4.**—A girl, age seven months, was brought to the clinic June 23, 1928. Since birth she had had a large mass in the sacrococcygeal region posteriorly. Roentgenograms revealed lower lumbar and sacral spina bifida. On June 28, 1928, the mass, which measured 25x15x10 cm., was removed from the sacrococcygeal region. A second mass, about 7x5 cm., and ventral to the sacrum, was later found and this also was removed. Recovery was complete. The pathologic diagnosis was dermoid cyst.

**Case 5.**—A woman, age 48, registered at the clinic September 23, 1929. She complained of distress in the lower part of her pelvis and rectum of six months' duration. Rectal examination revealed a fluctuating mass ventral to the sacrum, 2 cm. above the anal margin. Roentgenograms of the sacrum were negative. On September 27 the fluctuating mass was incised and about 100 cc. of thick pus escaped. The thick-walled sac was removed and recovery was complete. The pathologic diagnosis was dermoid cyst.

**Case 6.**—A woman, age 40, registered at the clinic December 5, 1932. She had had a dull pain in the lower part of her abdomen and sacral part of her back for 15 years. A perirectal draining sinus had been present since incision of an abscess years before. Rectal examination revealed a hard, irregularly lobulated mass arising from the left lateral wall of the sacrum. On December 8, 1932, the mass was incised and 120 cc. of putty-like matter escaped. Marked inflammation was present. The mass was excised as completely as possible. Slight drainage persisted intermittently for some time. The pathologic diagnosis was dermoid cyst.

**Case 7.**—A woman, age 44, registered at the clinic August 9, 1934. For three months she had noticed some discomfort in the rectum after a bowel movement. She had accidentally discovered a "tumor" in her rectum three months prior to registration while giving herself an enema. Rectal examination revealed a smooth soft mass ventral to the sacrum, and roentgenograms revealed spina bifida of the fifth lumbar and first sacral vertebrae. On August 21, 1934, a mass measuring about 7x5 cm. was enucleated from the ventral portion of the sacrum. Recovery was complete. The pathologic diagnosis was dermoid cyst.

**Case 8.**—A girl, age two and one-half years, registered at the clinic June 24, 1935. She had had a gradually enlarging tumor over the lower end of the sacrum since birth. Rectal examination revealed a mass measuring 4x8 cm., ventral to the sacrum and palpable just within the rectal sphincter. Roentgenograms revealed an anomaly of the last sacral vertebra. On June 28, 1935, a cystic mass filled with thick creamy material



was found extending from the coccyx to the ventral surface of the sacrum; it was completely removed. It measured 5x4x3 cm. Drainage persisted for some time. The pathologic diagnosis was dermoid cyst.

**Case 9.**—A woman, age 33, registered at the clinic July 6, 1936. She had had a dull constant ache in the lower part of her back for more than 15 years. Rectal examination revealed a mass 10x6x4 cm., lying ventral to the sacrum and to the left of midline. It was fixed and was not tender. Roentgenograms revealed a soft tissue shadow in the lower part of the pelvis. On July 10, 1936, the mass, which was found to be well encapsulated, was removed in its entirety. Recovery was complete. The pathologic diagnosis was dermoid cyst.

It is interesting that each of these nine patients was a female. Three of them were infants, and the average age of the other six was 36. The infants presented sacrococcygeal tumors at birth, two patients had had symptoms for 15 or more years, and four patients had had symptoms for from three to seven months. In two cases incision of an "abscess" had been performed; this was followed by a persistent discharge until the time of excision of the dermoid cyst. Roentgenograms were made in seven cases, and in two cases associated lumbar and sacral spina bifida was noted. In one case there was an anomalous last sacral vertebra.

The symptoms are not pointedly suggestive. Four patients complained of dull aching distress in the lower part of the back or abdomen, or in the rectum. One patient complained of a perirectal draining sinus, and one complained of partial urinary and rectal incontinence. The three infants presented definite evidence of a tumor. Rectal examination revealed the mass ventral to the sacrum in seven cases; rectal examination was not made in the cases of two infants. In those cases uncomplicated by a draining sinus or a previous operative attempt at drainage or removal, rectal examination revealed smooth fluctuant masses which were movable or only partially fixed and were not tender on pressure. In cases complicated by previous operation to institute drainage, marked inflammatory reaction may occur about the tumor and it may then become fixed and tender.

The posterior surgical approach is the one of choice. Complete removal of the lining of the dermoid cyst is essential and, usually, complete enucleation can be carried out without difficulty. If this is done the wound heals rapidly and complete relief of symptoms will follow.

**TERATOMATA.**—Teratomata are tumors in which tissue representing all three embryonal layers are arranged in disorderly fashion. Their origin is not well defined, various theories having been presented which are based on either their bigerminal or monogerminal origin. The bigerminal theory explains the origin of such tumors as an independent development of a blastomere during segmentation, a rudimentary duplication, or the so called parasitic twin. The monogerminal theory explains their origin as disorderly growths of remnants of fetal structures.

In the ventral sacrococcygeal region there are possibly sufficient fetal remnants to explain the various types of teratomata without resorting to the bigerminal theory of rudimentary twin. Middeldorpf was the first to attribute teratoma ventral to the sacrum to the persistence of a fetal structure. He pre-

sented the case of a girl, one year old, who had had a tumor in the region of the anus since birth. It opened to the outside and occasionally discharged a dark brown mucoid fluid. There was no connection with the rectum. This tumor was composed of fatty tissue containing a structure resembling a small loop of intestine. He believed that the tumor was derived from the postanal gut. Hansmann<sup>7</sup> was of the opinion that when the neurenteric canal remnant was the source of a tumor, there was frequently an associated anterior sacral defect.

Teratomata are most frequently found in the newborn and in infants. The attachment to the rectum is not intimate, but it may be to the sacrum or coccyx; they may be attached to the sacrum by a pedicle or may be enveloped in a capsule.

**Case 1.**—A girl, age seven, was brought to the clinic June 20, 1929. A large mass protruding from the perineal region had been noted since birth. Rectal examination was not made. Roentgenograms revealed a large soft tissue mass in the pelvis containing fragments of bone. The sacral curve was absent. At operation, June 25, 1929, the mass was found to be attached to the ventral median raphe of the sacrum and coccyx, displacing the genitalia downward and anterior. This mass was completely removed. It weighed 550 Gm. The perineal portion measured 10x7 cm. and the portion ventral to the sacrum was 9 cm. in diameter. Recovery was complete. The pathologic diagnosis was teratoma.

**MISCELLANEOUS TUMORS.**—This group of tumors ventral to the sacrum consisted of three malignant tumors (a fibrosarcoma, chondromyosarcoma, and a squamous cell epithelioma, possibly vesical in type) and one benign tumor (fibroma). There was no evidence of malignancy elsewhere in any of the cases of malignant tumor. Two of these patients with malignant lesions had symptoms of nerve root pressure; the third complained only of pain and constipation. In the case of the fibroma there was marked inflammatory fixation, which permitted only partial removal.

**Case 1.**—A man, age 51, registered at the clinic July 9, 1928. He had had a dull aching pain in the rectum, bearing down in type, of increasing severity for one year. Weakness in his legs had been present for six months, sciatica for two months, and urinary difficulty for two months. Rectal examination revealed a relaxed sphincter and an irregular, nontender mass ventral to the sacrum. Roentgenograms of the pelvis were negative. On July 18, 1928, a large tumor, 10x15x5 cm., was found eroding the ventral surface of the sacrum. The mass was removed almost in its entirety; it weighed 122 Gm. The patient died one year later. The pathologic diagnosis was fibrosarcoma.

**Case 2.**—A man, age 73, registered at the clinic November 29, 1934. He had had a mass in the coccygeal region with pain on sitting for eight months. Constipation had been present for six months. He had lost 35 pounds (15.9 Kg.) in the two years prior to his registration. Rectal examination revealed a firm mass ventral to the sacrococcygeal area, and roentgenograms revealed destruction of the lower part of the sacrum and coccyx. A soft tissue shadow was present. On December 12, 1934, the tumor, which had encroached on the rectum, was incompletely removed. It weighed 350 Gm. Again on January 14, 1936, a recurrent tumor about 6x4 cm. was removed. Radium treatment was instituted and there was some improvement. The pathologic diagnosis was chondromyxosarcoma.

**Case 3.**—A man, age 53, registered at the clinic December 26, 1931. He had had severe sciatic like pain, shooting in character, for four months. Some numbness and tingling of the left leg had also been present for four months. Rectal examination revealed a hard, fixed mass behind the rectum ventral to the sacrum. It was not tender. Roentgenograms of the pelvis were negative. At operation January 5, 1932, the mass,

which was found to be high in the hollow of the sacrum, was incompletely removed. Extensive radiotherapy followed. The patient died six months later. At the time of his examination and at operation no other evidence of malignancy was found. The pathologic diagnosis was squamous cell epithelioma, possibly vesical in type.

**Case 4.**—A woman, age 29, registered at the clinic February 15, 1929. She had had a dull aching pain, without projection, in the sacral region for six months. Rectal examination revealed a mass posterior to the rectum. The mass was rounded, smooth, not tender, and extended almost to the promontory of the sacrum. At operation, March 1, 1929, the mass was found to be quite fixed and edematous. It was incompletely removed. The pathologic diagnosis was fibroma.

## SUMMARY

Tumors arising ventral to the sacrum probably arise from remnants of fetal structures. The most important fetal changes in this region include the development and subsequent disappearance of the notochord, neurenteric canal, procotodeum and hindgut.

The subjective symptoms of tumor ventral to the sacrum are not definitely suggestive but include pain in the sacral region, constipation, and vesical and rectal incontinence. Rectal examination revealed the mass in each case in which it was made. It is the most important single diagnostic procedure.

The most satisfactory treatment of such tumors is surgical excision through a modified Kraske type of posterior incision. In the case of malignant tumors complete removal is often difficult and radiotherapy may help to relieve pain and to prolong life.

The incidence of recurrence is high, and recurrence is usually local. The prognosis varies with the type of tumor, being poor in instances of malignancy.

## REFERENCES

- <sup>1</sup> Chesky, V. E.: Sacrococcygeal Chordoma. *Arch. Surg.*, **24**, 1061-1067, June, 1932.
- <sup>2</sup> Cunningham, D. J.: *Textbook of Anatomy*. Ed. 5. New York, William Wood and Company, 27-55, 1923.
- <sup>3</sup> Ewing, James: *Neoplastic Diseases: A Treatise on Tumors*. Ed. 3. Philadelphia, W. B. Saunders Company, 1035, 1928.
- <sup>4</sup> Feldmann, I.: Chordoma Ossis Sacri. *Beitr. z. path. Anat. u. z. allg. Path.*, **48**, 630-634, 1910.
- <sup>5</sup> Fletcher, Eleanor M., Woltman, H. W., and Adson, A. W.: Sacrococcygeal Chordomas: a Clinical and Pathologic Study. *Arch. Neurol. and Psychiat.*, **33**, 283-299, February, 1935.
- <sup>6</sup> Galletly: Quoted by Raven, R. W.: Sacro-coccygeal Cysts and Tumors. *Brit. Jour. Surg.*, **23**, 337-361, October, 1935.
- <sup>7</sup> Hansmann, G. H.: A Congenital Cystic Tumor of the Neurenteric Canal: with Special Reference to Its Histology and Pathological Significance. *Surg., Gynec. & Obstet.*, **42**, 124-127, January, 1926.
- <sup>8</sup> Hundling, H. W.: Ventral Tumors of the Sacrum. *Surg., Gynec. & Obstet.*, **38**, 518-533, April, 1924.
- <sup>9</sup> Linck, A., and Warstat: Quoted by Kwartin, Boris, and Stewart, J. D.: Sacrococcygeal Chordoma. *ANNALS OF SURGERY*, **86**, 771-775, November, 1927.
- <sup>10</sup> Middeldorpf, K.: Zur Kenntniss der angeborenen Sacralgeschwülste. *Virchow's Arch. f. path. Anat. u. Physiol.*, **101**, 37-44, July, 1885.
- <sup>11</sup> Ribbert: Über die Ecchondrosis Physalifora Sphenooccipitalis. *Centralbl. f. allg. Path. u. path. Anat.*, **5**, 457-468, 1894.
- <sup>12</sup> Stewart: Quoted by Harnos, Oscar, and Palmer, L. A.: Chordomata and Report of Case. *Virginia Med. Month.*, **62**, 638-648, February, 1936.